

Endovascular management of dural fistulas into the cavernous sinus. A systematic review

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Abstract

Background: Dural fistula to the cavernous sinus (DFCS) is an infrequent pathology that consists in the anomalous communication between the meningeal branches of the internal carotid artery (ICA) and/or the external carotid artery (ECA) and the cavernous sinus.

Aim: To perform a systematic review to evaluate clinical and imaging findings in DFCS, and current indications for treatment.

Methods: A literature search was performed in several medical databases using the keywords "intracranial dural fistula", "carotid-cavernous fistula", "endovascular treatment", associated with "outcome"; resulting articles were assessed by considering factors such as: number of patients treated, type of material used, complications, and type of image technique used for diagnosis.

Results: 33 articles were selected, yielding: clinical and imaging: The symptoms are basically related to the type of venous drainage

of the fistula. The combination of ocular symptoms and tinnitus is highly suggestive of DFCS. Ocular symptoms are found in 80-97% of patients, while the tinnitus is present in up to 50% of cases. The imaging method for initial assessment of the DFCS is the magnetic resonance imaging. Digital subtraction angiography is the method of choice to determine adequately the precise angioarchitecture of the injury and its drainage. This data is of vital importance in future decision making. Treatment: Currently are considered as indications for the management of DFCS: 1) rapidly progressive deterioration of visual function, 2) angiographic evidence of abnormal cortical venous drainage, 3) the hypoxic consequences in retina and optic nerve, and 4) ischemic keratitis; the most suitable materials for embolization of the DFCS are CA and PAP.

Conclusion: DFCS stills being an uncommon cerebrovascular condition, with good outcomes from endovascular treatment.

Key words: Dural fistula; carotid-cavernous fistulas; N-butyl-cyanoacrylate; cavernous sinus.

Introduction

Intracranial arteriovenous fistulae are of two main types: carotid-cavernous fistulas (CCF) and dural arteriovenous fistulas. CCF is an infrequent pathology that consists in the anomalous communication between the meningeal branches of the internal carotid artery (ICA) and/or the external carotid artery (ECA) and the cavernous sinus. These belong to the indirect CCF. Unlike direct CCF which are frequently evocated by trauma, (1–5) indirect CCF more commonly occur spontaneously, being rare the traumatic etiology. (5, 6) Clinical manifestations generally follow a less severe course than direct CCF, being the ophthalmological symptoms and the presence of superior drainage, the most relevant data, mainly for provide an adequate therapeutic approach. Currently the preferred management is the endovascular. In this article we present a systematic review about endovascular management of dural fistulas into the cavernous sinus.

Methods

A literature search was performed in several medical databases including PubMed, EMBASE, Scopus, and Google Scholar; without restrictions or filters with regard to language or year of publication; showing a total of 33 results using the keywords “intracranial dural fistula”, “carotid-cavernous fistula”, “endovascular treatment”, associated with “outcome”. Emphasis was given to the

angiographic, imaging and hemodynamic characteristics found usually in dural fistulas to the cavernous sinus (DFCS); the resulting articles were assessed by considering factors such as: number of patients treated, type of material used, complications, and type of image technique used for diagnosis.

Etiology

Indirect CCF occurs typically and spontaneously in postmenopausal women, although they can occur also in other age groups as consequence of pregnancy, sinusitis or cavernous sinus thrombosis. (5–8). There is not a universal and convenient explanation regard its genesis, however, since Houser et al demonstrated that thrombosis of the lateral and sigmoid sinus precede the formation of dural fistulas, many authors implied the thrombosis of the cavernous sinus as the natural precursor of the DFCS, pushing away the theory proposed by Newton and Hoyt, who pointed out as cause, the microtraumatism over the delicate vessels of the carotid siphon (7).

Meyers et al (5), reported their 15 years' experience in the management of 135 consecutive patients with diagnosis of DFCS, being none of traumatic etiology. Therefore, the frank traumatic etiology of the DFCS is extremely rare. Berenstein et al (9) in 1986, reported 11 cases of arteriovenous fistulas, and mention one of DFCS of traumatic origin. Keltner et al (10) in 1987, described the case of traumatic indirect CCF within a total of 12 cases of CCF. Higashida et al (11) in an extensive review of 213 traumatic CCF published in 1989, found 7 cases of indirect fistula to the cavernous sinus. Jacobson et al

(6), in 1996 reported two additional cases of traumatic indirect CCF.

Notably, Jacobson and colleagues conclude that the recognition of traumatic origin allow an adequate choice of route and material to be used, we do not think that the traumatic history is a planning guide for management, we believe the choice of route and the material will be governed by the amount and type of tributaries that have the fistula, and venous drainage pattern, considerations that go far beyond agreement cited by other authors (6, 7, 10, 12).

Clinical and imaging findings

Clinical findings

The symptoms are basically related to the type of venous drainage of the fistula, therefore: 1) anterior drainage into the superior ophthalmic vein originate ophthalmic symptoms, for example, exophthalmos and chemosis; 2) posteroinferior drainage into the petrosal sinus, basilar plexus and pterygoid plexus will produce murmurs, and deficit of some cranial nerves; 3) subsequent drainage into the superior petrosal sinus originates murmur; 4) cortical reflux into the sphenoparietal sinus and upper middle cerebral vein may cause infarction and hemorrhage; and 5) deep drainage into the deep middle cerebral vein and the uncal vein can trigger hemorrhages (13).

These symptoms affecting intracranial and extracranial structures can be grouped into four patterns (14): a) orbital including chemosis, proptosis, eyelid edema and periorbital pain, b) cavernous with ptosis,

diplopia, anisocoria and ophthalmoplegia, c) ocular with decreased visual acuity or increased intraocular pressure (> 20 mmHg or interocular difference greater than 5 mmHg), severe eye pain, glaucoma, and retinal hemorrhage, and d) cerebral, including cerebral venous congestion in the basal ganglia, brain stem or cerebellum promoting presence of epilepsy and hemorrhage.

The combination of ocular symptoms and tinnitus is highly suggestive of DFCS. Ocular symptoms are found in 80-97% of patients, while the tinnitus is present in up to 50% of cases. These symptoms are usually less dramatic than direct CCF, and rarely are life-threatening. The flow of DFCS is usually much lower than the direct CCF, being the most important reason to present a less aggressive clinical spectrum, however there are reports of significant morbidity such as blindness and stroke (15,16). About 30 to 40% cases of indirect CCF present cerebral hemorrhage due to the rupture of dilated cerebral veins as a result of retrograde drainage.

When DFCS is clinically suspected, the patient must be carefully examined for confirmatory signs. In the presence of an eye with engorged vessels, conjunctival erythema or proptosis is mandatory to auscultate the skull and facial bones searching for murmurs. Usually the affected eye is ipsilateral to the fistula, however there are cases of bilateral symptoms due to the commitment of both cavernous sinuses because communication between them through intercavernous sinuses.

Imaging findings

The imaging method for initial assessment of the DFCS is the magnetic resonance

imaging (MRI). The findings include: 1) increased volume of the cavernous sinus; 2) loss of the interface between the cavernous sinus and the intracavernous portion of the ICA, 3) dilation of the superior ophthalmic vein (17, 18).

Digital subtraction angiography (DSA) is the method of choice to determine adequately the precise angioarchitecture of the injury and its drainage. This data is of vital importance in future decision making. Therefore, the angiographic evaluation must provide specific information regarding arterial tributaries, the precise site of shunt and the venous drainage pattern. In many cases this analysis can only be complete during the therapeutic phase. For this, it is recommended the angiographical evaluation in two phases: 1) diagnostic catheterization and 2) supraselective catheterization, which can be performed during treatment of the injury.

Some authors have emphasized the importance of non-invasive alternatives modalities such as helical CT-angiography (angio-CT) and magnetic resonance angiography (MRA), which is supposed to characterize the injury angioarchitecture (18). However, even with the visualization of afferent vessels and draining veins, DSA only guides in analyzing how the different compartments involved are interrelated, and helps the understanding of the hemodynamics of the lesion, enabling better therapeutic approach. Thus, the DSA still remains the gold standard for assessing DFCS.

Barrow et al (19) proposed an angiographic classification for CCF, revalidated by Debrun et al (7), and widely used by diverse authors,

see Table 1. In the review of literature persists the trending of the predominance of the CCF type D over type C, being truly extraordinary the type B.

TABLE 1
Barrow classification of DFCS

Type	Fistulous vessels	Characteristics
A	Carotid artery to cavernous sinus	Direct, high flow, the most common
B	Dural ICA branches to Cavernous sinus: -Meningohypophyseal trunk (66%) -Inferolateral trunk (30%)	Indirect, low flow
C	ECA branches to Cavernous sinus: -Internal maxillary (67%) -Middle meningeal (31%) -Ascending pharyngeal (24%)	Indirect, low flow
D	Both ICA and ECA branches	Indirect, the most common of low flow

ICA: internal carotid artery; ECA: external carotid artery

Why and when to treat DFCS?

From all dural fistulas, about 10% to 60% can present spontaneous resolution, and 25 to 50% are to the cavernous sinus, therefore have a much more benign course than direct CCF with a natural history that can lead to spontaneous healing. (10, 15, 16, 19–22) The cure occurring shortly after performing diagnostic angiography is not rare. Many patients experience a period of worsening of symptoms, particularly ophthalmics, during the spontaneous healing process. (12, 23) For this reason the decision on the management of the DFCS is controversial. Keltner, et al (10) treated by endovascular approach only 1 of the

10 cases reported, while 7 were offered only ophthalmologic management with treatment for glaucoma, reporting good control, therefore conclude that these cases should not be subjected to definitive treatment.

Currently are considered as indications for the management of DFCS: 1) rapidly progressive deterioration of visual function, 2) angiographic evidence of abnormal cortical venous drainage, 3) the hypoxic consequences in retina and optic nerve, and 4) ischemic keratitis (5, 11, 12, 20). Among the mechanisms involved in the worsening of dural fistulas are: 1) stenosis or thrombosis of the outflow, 2) increasing of the arterial blood flow, and 3) the emergence of a new fistula or enlargement of preexisting shunt (21, 24).

In the days before the advent of Neurological Endovascular Therapy, traditional surgery for the management of this condition was accompanied by a very high risk of thromboembolic events, almost always accompanied by cerebral ischemic disease and/or blindness (11). Currently the endovascular therapy is considered as the treatment of choice for all types of CCF for the following reasons: 1) is performed under local anesthesia with the possibility of ongoing clinical evaluation of the patient for early detection of any complication, 2) is a low risk approach, 3) less time consumption during the procedure, 4) reaches surgically inaccessible sites, 5) more likely to conserve the ICA blood flow, 6) fast recovery of the patient, and 7) there is always the possibility to repeat the procedure in case of initial failure or recurrence of the fistula (3–5, 7, 9, 11, 12, 20, 25)

Approach route

The DFCS are arteriovenous or arteriovenous shunts, in these cases it is determined that occlusion of the venous portion is the treatment of choice (26–28) for this purpose transvenous route is usually the best access method for insertion and release of embolic material, with current predominant use of detachable coils. Different routes have been described and used to reach the cavernous sinus, such as through lower petrosal sinus (contralateral or ipsilateral), basilar plexus, circular or intercavernous sinus, through the facial vein, the angular and superior ophthalmic veins or through the pterygoid plexus (5, 11, 20, 24, 25, 29–34).

In a large retrospective study, Mayers et al (5), summarizes 15 years of experience in the management of 135 DFCS patients treated transvenously, coils were used in 87% of cases, pieces of suture material in 13% and only in 3% of patients was used liquid adhesive material. They reported good long-term results in 97% of cases. Cheng et al (25) reported the cases of 27 patients treated transvenously with GDC and fibered coils, where only one case was necessary to combine with the arterial line for administration of PAP; in these same study, 89% of the cases experimented angiographic cure (25).

Retrospectively, Klish et al, reported 11 cases of DFCS embolized by transvenous approach with coils, reaching a frequency of anatomical cure of 63%, however, they refer that all patients, including those who did not achieve such healing, showed improvement in their symptoms (20). Reported complications of the transvenous approach with occlusion of

the cavernous sinus, the lesion of the vein access, the immediate and rapid increase of the ophthalmologic symptomatology and the injury of any of the oculomotor nerves caused either by over-packing of coils or induced thrombotic process itself within the sinus (10, 25). Furthermore, it has been determined in the case of dural fistulas that sinus therapeutic occlusion with coils, and the subsequent thrombosis can activate angiogenic factors within the adjacent dural walls (22, 32, 35–37).

In cases where it is not possible to exploit adequately the venous approach, the arterial approach can be useful (11, 12). CA is the material of choice in the arterial approach because allows better control of time and degree of polymerization modifying its dilution with the lipid contrast dye without starting with a total sacrifice of the circulation of the cavernous sinus, situation that makes it a more controllable material than PAP.

Some authors not use the transvenous approach just for the likelihood of these complications plus it is expensive for the number of coils typically required to occlude the cavernous sinus, fact that in economic stringency environments becomes a major limiting factor in the decision making.

One limitation of the arterial approach is the partial treatment of the fistula, particularly in type D cases, in which only were treated the fistulous pedicles of a single axis. This, in theory, perpetuates the fistula and may even increase the flow in other components, but our percentage of success and immediate and long-term cure is high. We think this is because the decision to embolize the pedicles of greater flow, and allow the passage of

embolic material to the malformation own nest which could induce a progressive thrombosis of the compromised segments of the cavernous sinus, this thrombosis is seen further promoted by combination with manual compression maneuver of the carotid artery. Hence can be avoided the nerve damage of oculomotor nerves or ophthalmologic deterioration that can be observed with the venous approach, and is a far less expensive method.

Satomi et al (24), have shown that partial treatment of DFCS, to what they have called palliative treatment, induces progressive changes in the pattern of venous drainage of the cavernous sinus with a trend towards very benign evolution with minimal ophthalmological commitment and almost no dilated cortical veins.

Another advantage observed with the arterial approach is the use of catheters guided by the flow, so far as possible avoids the use of microguides, which are to blame for vascular injury in case of venous approach. Among the complications of trans-arterial route, Viñuela et al (12) reported migration of the CA into distal branches of the middle cerebral artery, fortunately without much clinical implication, in addition to the transient increase in retro-ocular pain of one patient.

Embolization material

Viñuela et al (12) concluded that the most suitable materials for embolization of the DFCS are CA and PAP. The use of CA is currently widespread in the management of arteriovenous malformations (AVM), the opposite situation occurs in DFCS with few reports or small series. CA was first used as

management of direct CCF in a case treated by Bank et al (38) and in three cases managed by Kerber et al (39) reported in 1978 and 1979, respectively in which is highlighted the conservation of the carotid flow which was established in a situation really encouraging, in a time when most endovascular and surgical techniques aimed to blood flow occlusion of the ICA.

It is noteworthy that the experience of the last authors was when acrylic was not approved by the FDA for use in humans in northamerica. So was criticized and considered dangerous, and that the three cases described by Kerber et al (39) presented some type of deficit arising from the migration of the substance or by the application of the microcatheter or the non-detachable ballon technique used to administrate the substance, although the deficit was transient and the evolution of the patients was good in the short and medium term. Some authors like Pierot et al (40), Luo et al (3) and Wang et al (41) reported their successful experience with CA in management of direct CCF of traumatic origin, partially treated or recurrent. Berenstein et al (9) were first to use CA in the management of DFCS, which in turn was caused by trauma (9).

From 10 cases described by Keltner et al (10), only one was treated by endovascular technique with glue, precisely their single case secondary to trauma. Viñuela et al (12) described a series of 16 spontaneous DFCS, of which four were treated with AC, 1 with CA and surgery, 1 with CA and particles, and 3 with only particles. From all cases only one was identified as "inadequate" healing. These

authors conclude that the CA and the particles must be considered as the treatment of choice in the DFCS because allow superselective and more distal tributary vessels occlusion near the nest of the arterio-venous fistula, they consider that this may be sufficient to promote the cavernous sinus thrombosis with consequent total occlusion of the lesion.

Debrun et al (7) reported 132 cases of CCF, from which 32 were DFCS, of which 13 cases were treated only with CA, and the remaining with PAP or a mixture of both. Cure was obtained in 16 patients, whereas in the patients without early cure, some progressively improved, while others were lost to follow. It is necessary to emphasize that in this study the majority of indirect fistulas were type D (28 of 32 cases) and the authors conclude that they are the most difficult to treat because of the increased number of branches involved, which came from both ACE and ACI, hence adequate obliteration was more difficult to achieve, especially when considering that that occlusion of only ACE flow can result in increased participation of the meningeal branches of the ICA with much more difficult management in later stages (7).

Type D fistulas are more laborious, also require more mental exercise and can be disappointing for the endovascular specialist. For this reason some authors have considered to radiosurgery as a treatment option in selected cases, being combined with embolization or even with no pretreatment (42–46), however, more experience is needed with long-term outcomes to determine its efficacy and safety.

Conclusions

DFCS stills being an uncommon cerebrovascular condition, with good outcomes from endovascular treatment. Currently the endovascular therapy is considered as the treatment of choice for all types of CCF, because good outcomes, and because traditional surgery is accompanied by a very high risk of thromboembolic events, almost always accompanied by cerebral ischemic disease and/or blindness.

Abbreviations:

AVM: Arterio-venous malformation

CA: Cyanoacrylate.

CCF: Carotid-cavernous fistula.

DFCS: Dural fistula into the cavernous sinus

DSA: Digital subtraction angiography

ECA: External carotid artery.

GDC: Guglielmi detachable coils.

ICA: Internal carotid artery.

MRI: Magnetic resonance imaging

PAP: Polyvinyl-alcohol particles.

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